Professor Dyce
with the Author's
Compl.ts

OBSERVATIONS

ON THE

ARRESTED TWIN DEVELOPMENT

OF

JEAN BATTISTA DOS SANTOS,

BORN AT FARO IN PORTUGAL IN 1846.

(Illustrated by Woodcuts.)

BY

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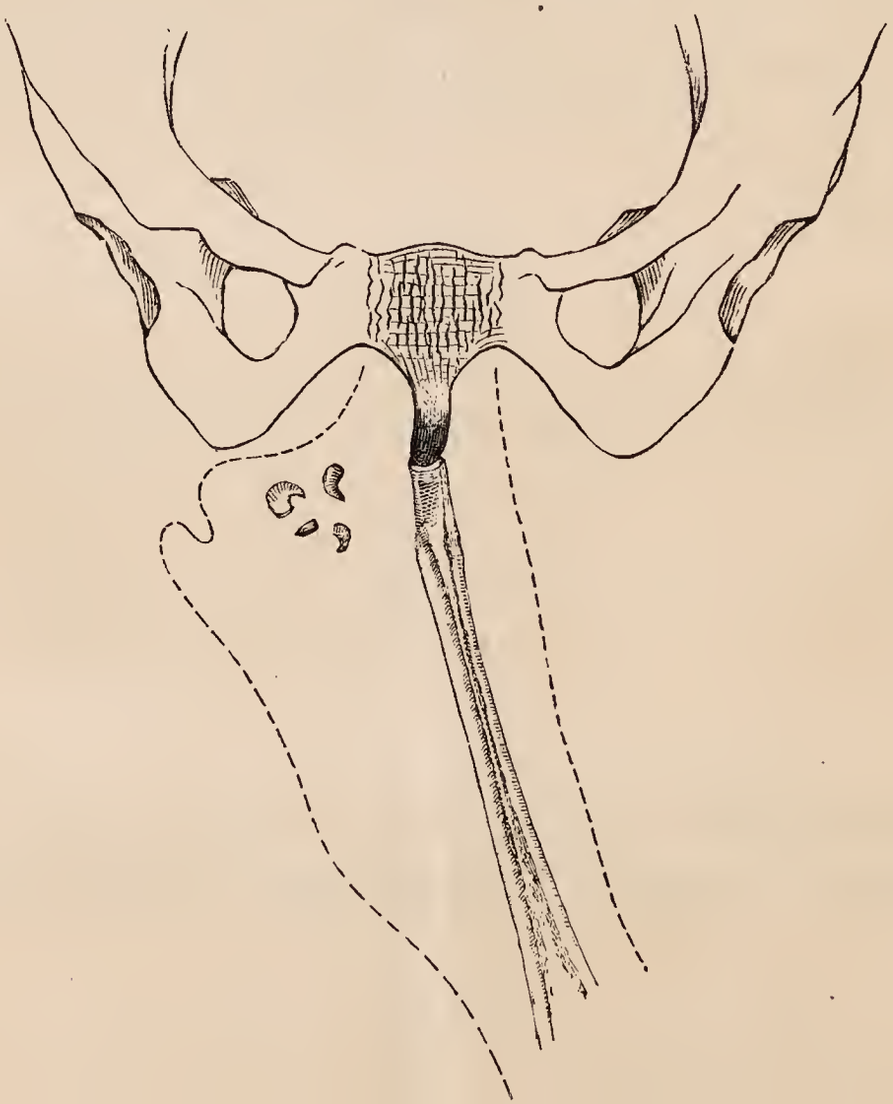

OBSERVATIONS

ON

ARRESTED TWIN DEVELOPMENT.

The profession is indebted to Mr William Acton, Surgeon to the Islington Dispensary,[1] to the Editor of "The Lancet," and to Mr Ernest Hart, of St Mary's Hospital, London,[2] for very interesting narratives of this remarkable double monstrosity in an adult. Attention having been thus directed to this perhaps unique variety of abnormity by excess, it may not be unimportant to record some anatomical peculiarities presented by it, yet hitherto unnoticed, so far as I am aware; and farther, I shall venture to express a difference in opinion relative to other features, on which the able authors of these two papers have made their observations.

When the subject of these remarks presented himself, on the 18th of December last, at the Royal College of Surgeons, Edinburgh, with his prominent and broadly cicatrized *umbilicus*, double *penis* and *scrotum*, and apparently double lower extremities, the idea suggested itself to me that from the umbilicus, possibly from the supra-sternal fossa, downwards, the existence of a twin production might be elicited.

I. *Abdominal Region.*—In seeking to trace development, as alone it can be done during life, the eye rests, *first*, on the broad base of depressed integument caused by the abnormal umbilical *raphé*. This wide mesial groove extends along the depressed *linea alba*, from the sunken ensiform cartilage, as low as the *pubes;* it is traceable, between the well-developed *recti* muscles, as a groove of above a quarter of an inch in depth, and almost four inches in breadth;

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[1] Mr Acton's interesting observations upon this case were made when Dos Santos was only six months old. *See* London Medico-Chirurgical Transactions, vol. xxix. pp. 103–106, with accompanying two characteristic drawings.

[2] That narrative, with four illustrative woodcuts, is published in the No. of that Journal for July 29, 1865.

and the edges of it begin, on a horizontal line with the lower edge of the umbilicus, to converge towards the pubic spines. The *umbilicus* is situated fully an inch nearer the pubes than is usual in the male sex; and it projects from its base, at the depressed *linea alba*, for half an inch forwards, so as to bulge a quarter of an inch in advance of the abdominal wall. Its base is two inches in diameter; while its sides and summit are irregularly nodulated, by means of two symmetrical depressions on its sides superiorly, and a horizontal groove on its lower half; and, withal, it is covered with a glistening stretched cicatrice, yet without any indication of umbilical hernia. This condition, then, of the parts, suggests the idea of the attachment to its summit, originally, of a double umbilical cord; and the photographic representation, prefixed to these remarks, may be observed to present the special features that I have now described. Upon coughing, no farther deficiency, nor any relaxation, are apparent in the abdominal or inguinal regions; neither, under this impulse, can any displacement, or vibration of bowel, be perceived on any part of their parietes, nor can more than two testicles be felt; and farther, on digging the fingers well into the hard and resisting *linea alba* in the hypogastric region during expiration, there cannot be discovered any abortive osteal element indicative of the lodgement, in the supra-pubic depression, of traces of the existence of a second pelvis.

II. *Chest, Neck, and Head.*—Proceeding to trace that mesial groove along the *sternum* to the neck, its depth is here fully one inch, its breadth nearly two inches, and its sides are rounded convexly forwards owing to the prominence of the cartilages of the true ribs. The *scrobiculus cordis* below, and the *jugulary fossa* above, are thus both absent; but there is no deficiency in the vertical conjunction of the osseous elements of the *sternum*. No abnormity is apparent on other parts of the neck, nor on the head or face. The thoracic, like the abdominal viscera, in relative position and size, appear normal.

III. *Spine.*—On examination of the head and trunk being made from behind, no deviation from the ordinary structure is observable. The mesial space between the *os coccygis* and the *anus* is depressed, deeply cleft, and somewhat tense; in a manner resembling the longitudinal mesial groove on the fore-part of the trunk.

IV. *Anus, Bladder, Urethra, etc.*—The orifice of the bowel is

situated unusually far forward, being about three inches anterior to the *os coccygis*, and immediately at the posterior edge of the point of attachment of the middle lower extremity. On introducing the forefinger as far as possible, and exploring slowly the parietes of the *rectum*, nothing unusual presents itself; and no information can thence be obtained as to the special arrangement of the parts contained within the *pelvis*. Refusal being still given to the proposal to sound the bladder *per urethram*, we are shut out from another source of information in regard to the anatomy of the *pelvis*. We have reason to conclude, however, from the answers given to inquiries put to Dos Santos, that he possesses a double bladder, the two communicating probably through an imperfect *septum;* that he has separate urethral muscles to each canal; and that he is furnished with only a single *levator ani*, and single vesical, vesicular, and prostatic plexuses of nerves and bloodvessels.

V. *Scrotum.*—The *scrotum* on each side is large, and its tissues seem natural. It is subdivided, by the usual *septum*, into two very unequal compartments; the external one on each side being large, and lodging a very large testicle of the usual form; while the internal, or mesial compartment of each *scrotum*, is small in size, and corrugated, though it is a quite distinct, pendulous, and flaccid bag or pouch. These latter are, respectively, of one inch and a quarter, and one inch and a half in length; and are stated to have lodged, in early life, two additional testicles, that, in boyhood, retired within the abdomen.

VI. *Penes.*—From the upper and fore-part of each *scrotum* hangs a *penis*, of large size, and of normal form. Each *penis* has, on its own side, precisely the same relations to the pelvic wall; the right *crus* of the right *penis* and the left *crus* of the left *penis* having, each, its usual origins, by *crura*, to the ascending *ramus* of *ischium* and descending *ramus* of *pubes;* the inner half of each *penis* belonging to the undeveloped twin. The mesial, or adjacent, *crura* of the two *penes* are equally well marked, though the incorporation and fusion together of the two mesial or intermediate *ossa pubis*, with their subjacent rami (including probably the adjacent *ossa ischii* and adjacent *acetabula* of the twin *pelves*), makes the tracing of them a little difficult. It seems, nevertheless, pretty clear that the origin of these intermediate *crura* is derived from a round thick fibro-cartilaginous process, which is found depending from the fore-part of the expanded pubic arch of the pelvis.

VII. *Interpubic descending process, representing the missing halves of a twin pelvis.*—This remarkable cartilaginous process presents to the touch a scabrous surface; it is about one inch in diameter, and three inches in length; and it is slightly incurvated backwards, ending inferiorly in a true arthrodial articular surface for the upper end of the united *ossa femorum.* The pelvic attachment of this fibro-cartilaginous process appears to be one of an amphiarthrodial nature, in connexion probably with the existence of a broader block than usual of the dense interpubic fibro-cartilaginous plates; and, in fact, to be an extension of these layers downwards, resembling the prolongation in tough cartilage of the extremity of a rib. The width of the pelvis, measured between the anterior superior iliac spines, and the very obscure yielding to torsion in either direction of this descending fibro-cartilage, appear to favour the view now advanced of the derivation and representative character of this remarkable structure.

Mr Hart's view of the "osteal congener of the true *penis*" being represented by the descending pelvic process just described, creates a necessity for the expression of my dissent from it, on the ground, that we cannot, in my opinion, be justified in concluding that in this dense process—*forming, as it does, an integrant part of Dos Santos' skeleton*—is to be recognised the rudiment of a *third* male organ. Moreover, Dr Sanders objects to Mr Hart's view because it would imply the existence of *a trin* development, of which there is no trace whatever.

This mesial projection, then, may be held to represent—situated, as it exactly is, in the place of the absent halves of the twin pelvis—the coalesced *ossa pubis*, including their proper ascending *rami* of *ischia* and *alæ* of *ilium.* In other words, this process may be regarded as the congener, and combination perhaps, at once of a broadened and elongated cartilaginous *symphysis*, the missing halves of the subpubic and triangular ligaments, and the flattened and prolonged surfaces of the *acetabula*, without, however, any apparent trace of the *teres* ligaments;—all these fused together to fulfil the double office the *ischium* so largely performs, that of affording a firm attachment to what in this case are the mesial *crura penis*, and the rudimentary two extremities to be presently described.[1]

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[1] The prefixed scheme or diagram may serve to illustrate this view:—A dotted outline represents the contour of the third limb; with a deep cicatrice, representing a rudimentary second *anus*, at its angular upper end.

VIII. *Perinæum.*—The *perinæum* proper is absent; the entire space between the anus and the posterior edge of the scrotum being occupied by the pendulous supernumerary limb.

IX. *Middle Lower Extremity.*—The superficial area of skin between the anal aperture and the two *scrota*, corresponding to the offset of this third limb, amounts to two and a-half inches in mesial length, and the space occupied transversely by its attachment is nearly two inches. The limb hangs loosely; is much bent at the knee; while the posterior surface of the thigh, and the entire leg and foot project forwards between Dos Santos' true limbs in such an inclination that the leg and foot rest in front of the inner surface of the lower two-thirds of his left thigh. This position of the abnormal limb—the most convenient one for Dos Santos in a sitting or lying posture—was probably impressed upon it *in utero*, and during infancy was apparently maintained as he lay on his back or side; while, subsequent to the age of progression, to avoid its unseemly protrusion, as well as the inconvenience of its loose dangling attachment above, he has been in the habit of wearing it slung or braced up firmly along his *right* thigh during the time he preserves the erect attitude.

The entire membral appendage is capable, without pain or strain, of very free rotation at its arthrodial attachment; sufficient indeed to permit of its being turned to either side about two-thirds upon its axis.[1] Under such free manipulation—more readily effected behind the person of Dos Santos whilst he stands erect—and with the forefinger inserted fully into the *rectum* for exploration, I satisfied myself that it is through the medium of the free arthrodial articulation, already referred to, that the middle extremity is suspended; and, moreover, that this articulation is a *congenital* formation, resembling, in structure and extent of mobility, the joint that exists between the lesser head of the *os humeri* and the shallow circular cup on the *radius*.

Projecting backwards from the posterior surface of the upper third of this middle limb, there is an obtuse enlargement of about four or five inches in elevation, directed upwards towards the cleft between the buttocks, and the skin covering the summit of which is remarkably depressed by an irregular cicatrice. On freely handling, and as it were kneading, the entire enlargement, it is found to

[1] The second and third figures in Mr Hart's paper express in part this extreme degree of mobility.

lodge, in the interspace between the cicatrice and the upper end of the *ossa femorum*, a congeries of about four separate and irregularly-shaped osteal elements, which may be taken to represent (as suggested to me by Dr Sanders) "the sacral bones" of the undeveloped twin—the elements probably of a second *os sacrum*. The value to the anatomist of the cicatrice alluded to may be regarded as great in determining the proper relation of parts, and important in the construction of any hypothesis on the uterine history of this double monstrosity. Consequently, in the absence of farther means of information than are at present available, it appears to me that in this irregular cicatrice we may recognise, as probably correct, the opinion which Mr Hart has advanced—in connexion with "the true anal aperture" being "deviated by about half-an-inch from the median line;"—namely, that "the puckered cul-de-sac in the skin over the clubbed and enlarged upper posterior extremity of the thigh is suggestive of an abortive second anus."

The *ossa femorum* I regard as plainly double, and fused together longitudinally. Their lower ends, laterally, present the well-marked contour and osseous markings of the normal flat external condyles. The *patellæ* are absent and wholly unrepresented. From the two *malleoli* bearing the elongated character of the lower ends of the *fibulæ*, I am satisfied that both of these bones, enclosing the *tibiæ* of both lower extremities, are well represented; and Mr Hart's second and third figures show admirably what this able surgeon so well describes as "really a coalescence of two feet, more or less perfect." No evidence, however, nor indication, of fracture, dislocation, false joint, torsion forwards, or other injury, are, in my opinion, capable of being detected along any part of the limb.

In testing the accuracy of the explanation that I have ventured to give,—that the contracted form and irregular disposition of the middle extremity have been imparted to it so as best to conduce to the comfort of the individual, an explanation which coincides also with the pathological state of the parts,—I would again recommend that an examination of these parts should be undertaken from a position *behind* the erect person of Dos Santos. Accordingly, the contracted abnormal limb should be rotated, at the subpubic arthrodial joint, so as to bring into view the back of the thigh upon the same vertical plane and parallel to the back of the two normal thighs. On this being done, we shall find little difficulty in immediately recognising the presence of contracted hamstrings, with contracted double tendon of Achilles, and double sole of foot; parts which sub-

tend the back of the abnormal thigh. We shall then see, in Dos Santos' additional limb, a marked instance of *congenital contraction of the hamstrings, combined with an extreme case of congenital Tales equinus.*

In viewing these features, it may be borne in mind that in all extreme cases of talipes equinus, owing to the shortened sural muscles and tendons,[1] the sole of the foot is so much extended at the ankle-joint that its plantar aspect is set at an obtuse angle with the calf of the leg. When, in addition to this, the flexor tendons of the knee-joint also are contracted, so that the region of the calf is set at an acute angle with the back of the thigh, then the lines of these two angles together seem to complete nearly three sides of a rhomboid, as on the margin; in which we find the sole of the foot running up parallel to the back of the thigh, and the dorsum of the retracted foot presenting its surface backwards,—that is, on the same vertical plane with the spinal column. Accordingly, on a close examination of Dos Santos, made while standing behind his person (as already suggested) a very acute angle—of 40°—may be noticed at his contracted knee-joint; an obtuse angle—of 158°—at his over-extended ankle-joint, as measured immediately over the insertion of the two united *tendines Achillis;* while his two contracted plantar *fasciæ*, united edge to edge, form, between the four well-marked eminences of the balls of the heel and great toes, one deep longitudinal grove or recess.

Apart from the remarks already made, presumptive evidence in favour of the *congenital* character of these contractions in Dos Santos' third limb is afforded, in the absence of every trace of the *patellæ;* bones which are not formed earlier than the third year of life.

X. *His general characters.*—It may be added, that Dos Santos' is an unusually interesting case of abnormal development, in so far as none of his functions, physical or mental, appear to be at all impaired; but are, if anything, rather above par. For his time of life, even with his nationality, he manifests a strong masculine development; and this is more especially visible not only in the structure of his skin, hair, muscular conformation—which is both robust and athletic,—breadth of shoulders, and strong marking of

[1] See a woodcut at page 11 of Dr Little's work on Club-foot, representing a preparation in the museum of the London Hospital, which exhibits the bones of a *congenital talipes equinus* in a young man. Here the plantar region of the entire foot forms *an obtuse angle with the calf of the leg.*

the bones; but likewise in his general manner, tone of voice, expression of countenance, mode of address, and perfect self-command.

General Remarks.

(*a.*) There seems reason to conclude that the foregoing case is *a twin, not a triple development;* the teratological law on this point, derived now from a very large induction, defining very clearly the very decided characters and extreme rarity of the latter form of monstrosity. Besides this, the well-marked and prolonged second perineal raphé, as described by Mr Hart, and in it the existence—as an exaggerated lacuna—of the rudimentary second *anus*, affords evidence that such a cul-de-sac could not have been the point of attachment of a third umbilical cord. Moreover, the two symmetrical and well-defined depressions upon the *umbilicus* proper, seem to point to the true seat of a probably double *funis*. And, farther, the all but invariable rule in regard to double monsters—that the union of dissimilar parts is very rare, and that like parts of two unequal bodies, the autosite and the parasite, are always attached to one another[1]—seems to make this case, as revealing such characteristics, speak for itself.

(*b.*) The *sternum* is abnormally broad, and deeply grooved, though not cleft; nevertheless its condition, as described, testifies to its being the compound of the lateral halves which in the normal state are fused into one bone. The apparently absent or much expanded *ensiform* portion denotes the same tendency to a separation in this region, at the mesial line. That this cleavage existed probably at an early period, the broad *linea alba*, cicatrice-like, appears likewise to affirm.[2]

(*c.*) There can be no doubt that the *two lower limbs* of the parasite, or secondary blighted fœtus, *are, in Dos Santos' case, fused into one.* Regnault's case of a somewhat analogous development[3] presents a twin formation, in which the two adjacent upper extremities coalesce, excepting only at the digits; and these are nine

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[1] See, for example, Plates 5, 14, 15, 16, 19, 20, of Isidore G. St Hilaire's Hist. Gen. et Part. des Anomalies, etc., tom. iii., which exhibit the congenital attachment of corresponding parts.

[2] In I. St Hilaire's *lib. cit.*, Pl. 14 (fig. 3), 16 (figs. 2 and 3), and 18 (figs. 2, 4, 5, 6), all these seven figures represent double monstrosity occurring at a cleft sternum,—the first six of these in man.

[3] St Hilaire, *lib. cit.*, Pl. 14 (fig. 1).

in number, the thumbs (like the great toes in Dos Santos' case) being united into one. J. F. Meckel[1] also, describes an intermediate third upper extremity (evidently coalesced) ascending in a straight line. Again, St Hilaire[2] represents one "symèle," in whom two lower limbs are fused together, ending in a row of eleven digits; another, presenting the bones of the legs and feet coalesced,[3] and terminating in eight digits, inverted, the little toes being unrepresented; and (at fig. 3), an interesting view, on dissection, of the disposition of the vascular trunks and the distribution of the nerves in this last case.

(*d.*) The only approximate type to this twin malformation—as regards a *third lower extremity consisting of two limbs fused into one, and having one sole of the foot with ten distinct digits*, and presenting some points of analogy as concerns *the position of the second anus and the umbilicus respectively*—which I find recorded, is one by St Hilaire.[4] Consideration of this case tends, I think, to support the views already expressed in the preceding narrative. But as regards Dos Santos' *sexual malformation*, we find no similar case of it, if we except the somewhat problematical histories cited and referred to by Meckel.[5] Six old writers, this author quotes, relative to so-called *double penes;* but I deem it relevant here to make one extract only:—"Illuc forsan pleraque exempla pertinent, quæ de pene vario modo duplici narrantur, præsertim ubi bini penes juxta se positi fuisse dicuntur. Hujus generis sunt, quæ citantur a Sinibaldo,[6] Hannæo,[7] aliisque minus integræ fidei medicis, quamvis per utrumque penem ad lubitum mejisse dicantur, in quibus hæc partium genitalium deformatio deprehendebatur et magnitudine solita uterque penis fuerit gavisus. Dissectio saltem talis difformitatis facta est nulla." The other four descriptions cited by Meckel, indicate, I think, merely varieties of *Epispadias* and *Hypospadias*.[8]

(*e.*) This case affords the nearest approximation to an instance of the "genre notomèle" of St Hilaire's "Monstres Doubles." In that malformation *an accessory limb is inserted in the region of the spine*,[9]

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[1] De Duplicitate Monstrosa Commentarius, Halae, 1815, p. 79.

[2] *Lib. cit.*, Pl. 5 (fig. 1.)

[3] *Lib. cit.*, Pl. 5 (figs 2, 3.) [4] Ibid., Pl. 20 (fig. 1.) [5] *Lib. cit.*, p. 50.

[6] Geneanthropia, lib. ii. c. 3. Ni autem totus fallor, vesica hic adfuit inversa. [7] Obs. anat. rar. cent. iv. hist. 22.

[8] J. F. Meckel, Handbuch der Path. Anat. Leipsig, 1812, i. 650–657.

[9] Tom. iii. p. 270.

and, as is stated by him, "est encore une monstrosité inconnue chez l'homme, et très rare chez les animaux." That Dos Santos' case supplies this desideratum in the human species, I do not mean to assert, unless it be assumed that the four osteal elements in his case are the elements of *a second sacrum and spine.*

(*f.*) A *somewhat similar parasitic mesial bone* to that of the middle lower extremity here described—being of a *tibia*-like form, covered with periosteum, and surrounded by a number of vessels and nerves—was found by Professor Sir James Simpson[1] within a tumour removed by Dr Richardson, of Stockton-on-Tees, "from over the middle of the *sacrum* of a child, without showing any bad symptoms afterwards."

(*g.*) The record of numerous instances of *contracted knees and clubbed-feet monsters* tells in favour of the view I have ventured to impress,—namely, that there lacks evidence to show that any change has come over the form and position of the third limb of Dos Santos, since his birth.[2]

(*h.*) Dos Santos' *abnormal limb exhibits little vascularity, sensibility, or voluntary movement;* it is surrounded at its neck of attachment to the perinæum by flaccid skin, loose fat, and connective tissue merely; nevertheless, the probability of there existing an important communication with the spinal canal, and the fact that *its sub-pubic connexions* are significant, forbid the notion that this redundant middle limb might be safely removed.

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[1] See his Lecture in the Medical Times and Gazette for July 2, 1859, pp. 5, 6.
[2] St Hilaire, *op. cit.*, Pl. 12 (figs. 1, 2, and 3.)

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